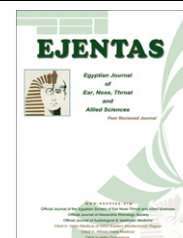




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CASE REPORT

Maxillary sinus arteriovenous malformation: A rare clinical entity

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Abstract Arteriovenous malformation (AVM) of the head and neck is rare but a life threatening benign condition. This paper reports a case of arteriovenous malformation located in the maxillary sinus, in a 16-year-old girl. Her main complaints were nasal obstruction and rhinorrhea. Computed tomography (CT) scan and magnetic resonance imaging (MRI) evoked this entity. Complete tumor removal was performed by the Degloving approach without significant bleeding. The clinical features, therapeutic approaches and the differential diagnosis are discussed.

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1. Introduction

AVM represents a direct connection between the arterial and the venous systems.^{1,2} It is commonly seen in the head and neck region which is endowed with an abundant and redundant arterial vascular supply. However, its emergence in the paranasal sinus is exceptional. The symptoms and radiographic appearances are highly variable. They may often indicate a benign condition but can induce a life-threatening hemorrhage.⁴

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2. Case report

A 16-year-old girl presented with a 6-month history of left nasal obstruction and intermittent smell foul rhinorrhea, without epistaxis, headache, diplopia, or sensory changes. She had no history of facial trauma. Endoscopic examination revealed a polypoid mass in the left nasal cavity. Examination of the right nasal cavity was normal, as was the remainder of the head and neck examination. Biopsy of the nasal mass was attempted but was limited by an abundant epistaxis.

Computed tomography (CT) with contrast showed a heterogeneously enhancing mass within the left nasal cavity and maxillary sinus. CT also demonstrated dilated vessels within the tumors on the arterial phase and enlargement of the internal maxillary artery (Figs. 1 and 2) Magnetic resonance imaging (MRI) revealed a heterogeneous mass extending from the nasal cavity into the left maxillary sinus, enlarging osteomeatal complex with destruction of the middle concha and the inferior



Figure 1 Coronal CT scan: arterial phase on contrast-enhanced CT demonstrated dilated vessels within the mass (white arrow-heads) and left maxillary artery enlargement (white arrow) compared to the right maxillary artery (black arrow).



Figure 2 Coronal CT scan: arterial phase on contrast-enhanced CT demonstrated dilated vessels within the mass (white arrow-heads) and left maxillary artery enlargement (white arrow) compared to the right maxillary artery (black arrow).

turbine bone. The lesion had intermediate signal intensity on T2 spin-echo sequence with intense enhancement after gadolinium injection and contained multiple serpentine areas of flow voids corresponding to high flowing blood in vascular channels (Figs. 3 and 4). The diagnosis of AVM was highly evoked. The



Figure 3 Coronal T2 MRI: heterogeneous poorly margined heterogeneous mass in the extending from the nasal cavity into the left maxillary sinus enlarging the osteo meatal complex with destruction of the middle concha and the inferior turbine bone. Intratumoral signal voids, which corresponded to high flowing blood in vascular channels (white Arrows). Obstructed secretions on the maxillary sinus and ethmoid cells were hyperintense compared to the mass (black arrows).



Figure 4 Coronal T1 MRI GADO(+) with fat saturation: avidly enhancing mass in the left nasal cavity extending to the maxillary sinus. Note the flow voids on intense hyposignal (black arrows). Trapped secretions within the sinuses are hypo intense compared to the mass with peripheral mucosal enhancement noted in maxillary sinuses (white arrows).

patient had undergone a surgery via the Degloving approach. Complete tumor removal was performed without significant bleeding. During 9 months follow-up, no complaints were reported.

3. Discussion

AVM is a common vascular tumor of the head and neck region. 31% of them are localized in the maxillofacial skeleton. The nasosinus localization is exceptional. To our knowledge, only two cases of maxillary sinus AVMs have been described.^{4,6}

AVMs can be acquired following trauma. But a majority is congenital arising as a result of aberrant vessel angiogenesis. They are commonly present at birth, asymptomatic until the occurrence of an additional growth or a vascular engorgement secondary to thrombosis, trauma, infection, puberty or pregnancy.

AVMs are usually diagnosed at adolescence. The mean age at presentation is 19 years without sex predilection.⁷

Clinical presentation of AVMs is not specific and subtle.⁸ The most common complaints are chronic intermittent epistaxis and nasal stuffiness. Further symptoms are: recurrent infections, headache, facial pain, facial asymmetry, facial discoloration, tinnitus, proptosis, ocular pain, toothache and teeth loosening with a "boggy" toothbed.^{3,8} Large lesions may be associated with high cardiac failure. Some lesions may result in consumption coagulopathy, requiring the transfusion therapy.⁸ Our patient had never complained from epistaxis or headache. Her main complaints were nasal obstruction and recurrent sinusitis.

Diagnostic is usually easy when endoscopic examination shows a pulsatile expansive nasal tumor.⁴ Atypical presentation had been, yet reported. On endoscopic examination, a nasopharyngeal AVM had been described as an adenoid vegetation.⁷

Imaging exploration is crucial to assess the diagnosis of AVMs, to evaluate the extent of disease and to discard differential diagnosis. Arteriography used to be the exploration of choice to confirm the diagnosis of AVM by delineating internal angio-architecture and depicting its three components: enlarged arteries, nidus and draining veins.⁹ However, it is an invasive procedure with a small but definite risk of stroke. Thus, conventional angiography is usually reserved for pretherapeutic evaluation.¹⁰

MR imaging (MRI) can be used for both diagnosis and follow-up of AVMs after therapy.⁷ On MRI, AVMs are characterized on spin-echo sequences by serpentine flow voids, indicative of the high flow rate of these lesions.^{7,10} MRI is useful in differentiating low and high-flow vascular lesions and delineating their anatomical extent allowing better planning of therapy.¹⁰ However, delimitation of the afferent and efferent dilated vessels is often difficult. MR angiography (MRA) demonstrates major feeding vessels and multiple intra-lesional vessels in relation to the high flow lesions. However, small feeding vessels to the AVM are not clearly identified.

Although 2-D TOF MRA can distinguish AVM from venous malformations, it cannot compete with catheter angiography for the identification of AVM feeding vessels.¹⁰

The usefulness of CT is limited. Application of newer CT techniques (multidetector helical CT with image reconstruction) may increase the role of CT in high-flow lesions, but the greater soft tissue contrast sensitivity of MR imaging remains its strong suit.¹⁰

Maxillary sinus AVM must be treated to relieve symptoms, and to avoid complications consisting in potential life-threatening hemorrhage, alteration of the normal structure

and function of the paranasal sinuses, maxillary dental arch, and nasal cavity.³

The management of AVM of the maxillary sinus and nasal cavity is complex and requires a multi disciplinary approach including surgeons and interventional radiologists. The main objective of the treatment is complete resection of AVM with the minimum bleeding risk.

Endovascular subselective embolization is indicated for occlusion of no resectable lesions, and preparation for surgical resection. It has become a widely used modality in the treatment of vascular abnormalities. However, possible complications such as occlusion of the underlying artery, ischemic stroke and autolysis of the embolization material over time should be considered.¹⁰

Surgery remains the treatment of choice of AVM.^{7,8} It should be carried out within 24–48 h, after embolization, to avoid rapid recruitment of collateral vessels.⁸ Several important points should be considered to achieve complete cure of this condition. The nidus of AVM should be completely removed. Ligation of the feeding artery without total removal of nidus should be avoided because it may enhance the growth of other quiescent feeding vessels uncontrollable by embolization.^{9,10} Incomplete resection could result in a larger lesion in a short period of time.

In our case, embolization had not been available. The Degloving approach permitted a total control of the nasal and maxillary cavities and complete removal of the tumor without notable bleeding.

Close long-term follow-up is needed. Angio MR may be repeated at 3–6 month intervals, following resection to detect early signs of recurrence.

4. Conclusion

AVM of the nasal vestibule and maxillary sinus is a rare benign condition but it may lead to a life-threatening hemorrhage requiring emergency endovascular or surgical treatment. Treatments include embolization, surgical resection, or a combined approach. Post operative long term close follow up is required to detect recurrence.

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